The Royal College of Surgeons of England

Ann R Coll Surg England 1999; **81**: 205–206

Case report

Adenocarcinoma within a rectal duplication cyst: case report and literature review

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Intestinal duplications are uncommon but recognised developmental anomalies. Duplications of the rectum are the most uncommon of these anomalies. They may present with perianal fistulae, bleeding, a pelvic mass or symptoms produced by a mass, or, rarely, malignant change. We present a case of an adenocarcinoma within a rectal duplication cyst which was initially thought to be inoperable but was treated by radical surgery.

Key words: Adenocarcinoma – Rectal duplication cyst

Case report

A 65-year-old retired housewife presented with a 3 month history of rectal bleeding, 2 years of pink rectal discharge, peri-anal pain and perineal parasthesia. She had suffered recurrent peri-anal sepsis and fistulae between the ages of 21 and 26 years. On digital examination, there was roughness of the posterior anal canal and rectum overlying a fixed extramucosal mass. Sigmoidoscopy showed no mucosal abnormalities. A CT scan demonstrated a mass extending from the anal canal to the lower sacrum and into the right ischiorectal fossa.

It was initially thought that the diagnosis was carcinoma of the anal canal with extramucosal spread but Trucut biopsy identified adenocarcinoma. The tumour was thought to be inoperable so the patient was treated with chemoradiotherapy. Six months after completion of treatment, the patient felt well but still complained of rectal bleeding.

The patient was then referred to St Mark's Hospital for consideration of radical surgery. On digital examination, there was a firm rubbery mass immediately above the pelvic floor in the posterior midline. This was fixed to the posterior wall of the rectum, and to the coccyx and lower sacrum. The mucosa of the rectum felt normal and mobile over the mass; sigmoidoscopy was unremarkable. She was admitted for further investigations. An abdominopelvic CT scan with contrast identified a 2×4 cm cystic mass anterior to the sacrum displacing the rectum anteriorly (Fig. 1). It was fixed to the sacrum but free from the pelvic side walls. These findings supported the diagnosis of a rectal duplication cyst.

It was decided to attempt cure by radical surgery. Initially, the patient was positioned supine and the abdominal component of a radical rectal excision was performed down to a level above the tumour, after which a colostomy was fashioned and the abdomen

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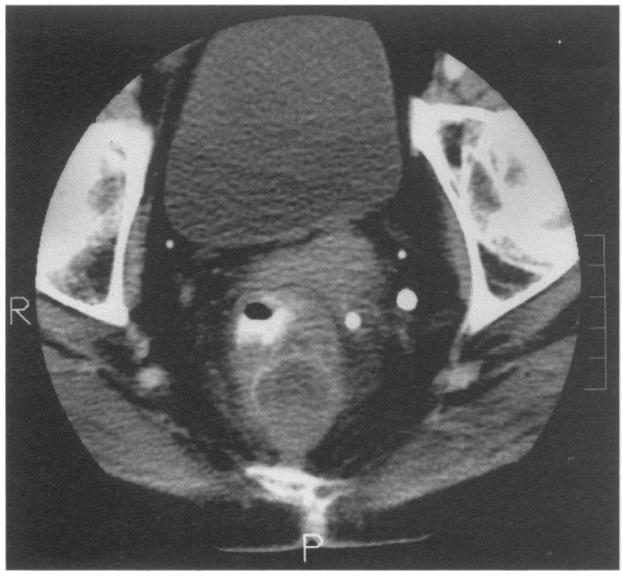

Figure 1 A CT scan with contrast showing a presacral rectal duplication cyst displacing the rectum (containing contrast) anteriorly.

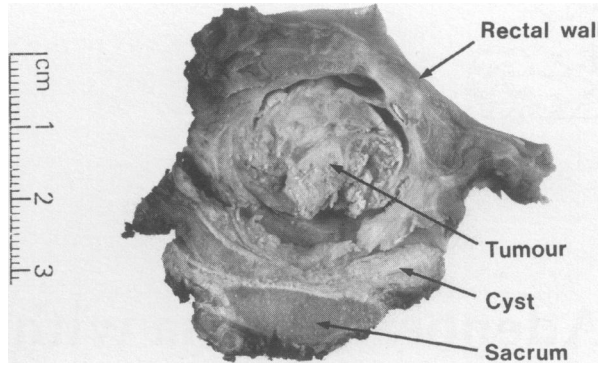

Figure 2 Transverse section showing a duplication cyst containing an adenocarcinoma with central necrosis. The tumour was infiltrating through the cyst wall, composed of smooth muscle of anal sphincter type which was adherent to the rectal wall.

closed. For the second stage of the procedure, the patient was placed in the prone jack-knife position; the rectum was mobilised from below via a circumferential anal incision which was extended cranially in the mid-line to allow partial excision of the sacrum, which was divided below the third sacral vertebra allowing *en bloc* removal. Postoperatively, the patient recovered well with no evidence of neurological sequelae.

Pathological examination confirmed that this was a an adenocarcinoma arising within a 'true' rectal duplication cyst (Fig. 2).

Discussion

Intestinal duplications are congenital cystic lesions which resemble, and are associated with, part of the gastrointestinal tract. They may occur in any region of the gut, but the rectum is the least common site. The most widely accepted aetiological explanation is that duplication occurs due to pinching off of a diverticulum present in the 8–9-week-old embryo.[1]

Rectal duplication cysts occur in the presacral plane and may present because of mass effects (pain, obstruction or urinary symptoms), fistulation or infection. Malignant change is extremely rare and was first described in 1932 in a 38-year-old woman.[2] In a review of the world literature, 24 cases of rectal duplication cyst were described of which only two contained adenocarcinoma.[3] Our review of the literature identified a further four cases of adenocarcinoma in a true rectal duplication.[4,5]

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Received 7 October 1998